

*Case Report*

## Twin pregnancy with a living fetus and coexisting complete hydatidiform mole

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**CASE REPORT** A 26 year-old para 2 presented at 30 weeks' gestation with a small, painless antepartum haemorrhage. When booked at sixteen weeks' amenorrhoea ultrasound scanning confirmed her dates to be correct and no obvious fetal abnormality was seen. The pregnancy was uncomplicated and the patient was well and normotensive throughout. All of her pregnancies were conceived with the aid of clomiphene citrate and her other children were delivered vaginally at term.

On admission the patient's blood pressure was 115/58 mmHg. The fundal height was at the xiphisternum which was greater than expected and the uterus was soft and non-tender. There was a single fetus, the lie was longitudinal, the presentation was breech and the fetal heart was heard. On ultrasound scanning the liquor volume was normal and the placenta was posterior and slightly low-lying. A large mass of multiple sonolucent areas was visualised separate from the superior edge of the placenta. Intravenous access was established and blood was taken for haemoglobin, group and hold (B Rhesus positive) and human chorionic gonadotrophin (HCG) level. Intramuscular steroids were given to the patient to improve fetal lung maturity.

Four hours after admission the patient experienced a sudden, substantial, painless haemorrhage. A second intravenous line was inserted, blood was taken for coagulation profile and 500 mls colloid was infused over ten minutes while four units of packed cells were awaited. On examination she was pale but normotensive. The uterus was soft and non-tender, the fetal heart rate was 130 beats per minute and fresh blood was seen trickling from the vagina. Written consent for an emergency caesarean section was obtained from the patient.

An emergency lower segment caesarean section was performed under general anaesthetic and a male infant weighing 1410 g was delivered in good condition by breech extraction. A healthy placenta and membranes were delivered, and in addition 1200 ml of vesicular tissue were removed from the uterus. An infusion of oxytocin was commenced and carboprost 250 µg was injected into the myometrium to improve uterine tone and control blood loss.

The patient made an uneventful recovery and went home on the fifth postoperative day. The baby remained in the neonatal unit and made very good progress. The BHCG result from admission was 251,478 IU/I and the pathology of the vesicular tissue was reported as a complete mole co-existing with a normal twin fetus and placenta. The patient's details were registered with Charing Cross Hospital in London for further follow-up and treatment. Her BHCG was normal within 8 weeks of evacuation of her uterus (urine level 18IU/I, serum level 4IU/I [normal values: urine 0-24IU/I; serum 0-4IU/I]). Follow-up will be for 6 months with monthly urine samples being sent to Charing Cross. The patient has not met any of the criteria to commence chemotherapy to date.

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## DISCUSSION

A twin pregnancy consisting of a complete mole and one normal fetus and placenta is extremely rare. An incidence of only 1/22,000 - 100,000 has been reported.<sup>1,2</sup> In the literature 43 cases have been reported since 1977 and from those only 13 infants have survived.<sup>3</sup> It has been suggested that the incidence of this condition may increase in the future due to the use of ovulation – inducing agents because of the increased multiple pregnancy rates associated with their use.<sup>2</sup>

Hydatidiform moles are abnormalities of placental tissue that involve trophoblastic proliferation and hydropic degeneration with absence of vasculature and are classified as being either complete or partial. The karyotype of a complete mole is usually 46XX with both chromosomes being paternal in origin. This occurs either by the fertilisation of an 'empty' egg by two sperm or by the doubling of the paternal 23X inside the egg.<sup>4</sup> A partial mole arises by the fertilisation of an egg by two sperm to give rise to a triploid conception and, therefore, even though a fetus is often present it is often abnormal.<sup>5</sup> It is important in cases of 'twin' molar pregnancies to make the distinction between a partial mole, which consists of a triploid fetus and placenta with hydatidiform changes and that of a normal fetus with a coexisting complete mole. This is because the evolution of a partial mole into choriocarcinoma has not yet been recorded in the literature.<sup>6</sup> In contrast, the risk for development of choriocarcinoma with a single complete mole has been reported as 14% and as significantly higher for twin pregnancies with complete moles (55%).<sup>7</sup>

Clinically, the patient may present with complications such as vaginal bleeding, severe pre-eclampsia prior to 20 weeks, hyperemesis gravidarum and hyperthyroidism. Vaginal bleeding is usually intermittent and variable in amount and vesicular material may even be passed vaginally. The uterine size may be large for dates as it was in this case. It is also of interest that the patient reported here conceived with the aid of clomiphene citrate predisposing her to a twin pregnancy.

The diagnosis of coexisting complete mole and normal 'twin' fetus can be made by assessing the clinical picture, measuring human chorionic gonadotrophin levels and abdominal ultrasound.<sup>4</sup> The diagnosis of complete mole can be difficult

in these circumstances because it is often not suspected when a normal fetus is seen on ultrasound. Human chorionic gonadotrophin levels are usually much higher than in a normal pregnancy but a molar pregnancy can produce normal levels and HCG may be elevated in a normal pregnancy.<sup>4</sup> Ultrasound can be useful in the diagnosis of abnormal placental tissue, with the classic appearance of a complete mole being described as resembling a 'snowstorm'. With a coexisting twin it can be more difficult to make the diagnosis on ultrasound especially in the first trimester but Steller<sup>7</sup> found that 68% of twin moles were diagnosed correctly by scanning.

The optimal management of this condition is not known because of its rarity and because most of the evidence in the literature is from single case reports.<sup>1</sup> The management depends to a great extent on the gestation at which the diagnosis is made. Before intact fetal survival following delivery has become a realistic probability (<28 weeks) the patient's health is the priority and the pregnancy is usually terminated.<sup>4,8,9</sup> Once the diagnosis is suspected the mother should be cautioned with regards to the potential risks associated with the continuation of the pregnancy namely pre-eclampsia, antepartum haemorrhage and hyperthyroidism. In addition she should be advised that the risk of persistent trophoblastic disease is significantly higher in twin pregnancies with a hydatidiform mole than for single molar gestations and that subsequent chemotherapy is also required for 57% of 'twin' moles compared to 19% of single moles.<sup>1,4</sup> Should a patient opt to continue the pregnancy she should be made aware that there is no accurate way of predicting invasive growth and she would require very close monitoring throughout the pregnancy.

Controversy remains regarding the management of cases where a normal fetus coexists with a molar pregnancy and many still advocate termination of pregnancy if an early diagnosis is made. In the present case this difficult decision did not have to be taken because the diagnosis was made at 31 weeks when the chances of fetal viability were realistic. Continuation of the pregnancy beyond this stage was not possible because the quantity of vaginal bleeding necessitated urgent delivery. The lack of ultrasound diagnosis in this case until 31 weeks illustrates that the presence of a normal looking fetus does not rule out the possibility of trophoblastic abnormalities.

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